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Peer Relationships in Children with Williams Syndrome: Parent and Teacher Insights

Amanda E. Gillooly[1], Deborah M. Riby[2], Kevin Durkin[3], Sinéad M. Rhodes[4]

Institute of Health and Wellbeing, University of Glasgow, Gartnaval Royal Hospital, 1055 Great Western Road, Glasgow, G12 0XH, Scotland, UK[1]

Department of Psychology, Durham University, South Road, Durham, DH1 3LE, England, UK[2]

School of Psychological Sciences and Health, University of Strathclyde, 40 George Street, G1 1QE, Scotland, UK[3]

Salvesen Mindroom Research Centre, Child Life & Health, Centre for Clinical Brain Sciences, University of Edinburgh, 20 Sylvan Place, Edinburgh, EH9 IUW, Scotland, UK[4]

Abstract

Although children with Williams syndrome (WS) are reported to show a strong motivation towards social interaction, evidence suggests many experience difficulties with peer relations. Less is known regarding the characteristics of such difficulties. Parents and teachers of 21 children with WS (7- to 16- years) completed questionnaires measuring aspects of social functioning and peer interactions. Parents and teachers reported that children with WS demonstrated significantly greater peer problems than population norms, including difficulties sustaining friendships and increased social exclusion. More substantial social functioning difficulties were associated with greater peer relation problems. The study provides multi-informant evidence of peer relationship difficulties in children with WS that require further consideration within the broader WS social phenotype.

Keywords: Williams syndrome, peer relationships, social functioning, social skills

Corresponding author:
Dr Sinéad M. Rhodes
sinead.rhodes@ed.ac.uk
0131 536 0642

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Abstract

Although children with Williams syndrome (WS) are reported to show a strong motivation towards social interaction, evidence suggests many experience difficulties with peer relations. Less is known regarding the characteristics of such difficulties. Parents and teachers of 21 children with WS (7- to 16- years) completed questionnaires measuring aspects of social functioning and peer interactions. Parents and teachers reported that children with WS demonstrated significantly greater peer problems than population norms, including difficulties sustaining friendships and increased social exclusion. More substantial social functioning difficulties were associated with greater peer relation problems. The study provides multi-informant evidence of peer relationship difficulties in children with WS that require further consideration within the broader WS social phenotype.

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Williams syndrome (WS) is a genetic disorder that arises from the hemideletion of approximately 28 genes from one copy of chromosome 7q.11.23 (Haas et al., 2013). WS has an estimated prevalence between 1 in 7,500 and 1 in 20,000 (Sparaci, Stefanini, Marotta, Vicari, & Rizzolatti, 2012) and is characterised by mild to moderate levels of intellectual impairment (Searcy et al., 2004) a profile of cognitive strengths and limitations (Bellugi, Adolphs, Cassady, & Chiles, 1999), medical complications which include cardiac difficulties (Järvinen-Pasley et al., 2008), and an atypical social phenotype (Jawaid et al., 2012). Relating to the latter of these, many WS individuals are often reported to show a hyper-social profile (Frigerio et al., 2006; Jones et al., 2000), with characteristic traits including a strong predisposition to engage in indiscriminate social interactions (Lough, Rodgers, Janes, Little, & Riby, 2016), reduced 'stranger danger' awareness (Riby, Kirk, Hanley, & Riby, 2014) and subsequently increased social vulnerability (Riby, Ridley, Lough & Hanley, 2017). While the social drive suggests that individuals with WS are socially motivated, this hyper-sociability occurs alongside difficulties with complex social cognition and social communication (Klein-Tasman, Li-barber, & Magargee, 2011).

During social interactions, children with WS often demonstrate atypicalities of social functioning (Klein-Tasman et al., 2011) that can include (but are not restricted to) difficulties with social communication due to their reliance on stereotyped speech (Laws & Bishop, 2004; Udwin, Yule & Martin, 1987), and difficulties interpreting subtle social cues from others (Klein-Tasman, Mervis, Lord, & Phillips, 2007; Porter, Coltheart, & Langdon, 2008, Tager-Flusberg & Sullivan, 2000).These atypicalities of social functioning, in combination with the children's mild to moderate levels of intellectual impairment can be suggested to contribute to increased levels of social vulnerability, as highlighted by a characteristic 'hypersocial' profile. The children's intellectual disabilities are also suggested to make establishing friendships with peers substantially more challenging, particularly when

differences in interest and abilities are more prominent. These atypicalities are reported to transcend across social contexts and across ages. Klein-Tasman et al. (2011) obtained parents' and teachers' ratings for 4-16-year-old children with WS on the Social Responsiveness Scale (SRS; Constantino & Gruber, 2012) and found substantial difficulties across several components of social functioning, but in particular for social cognition and social communication. Only 13% of the children with WS scored within the 'normal' range for overall social functioning. These results have been replicated by Van der Fluit, Gaffrey and Klein-Tasman (2012) and Riby et al. (2014). Taken together, the evidence to date provides support for a profile of substantial atypicalities of social functioning.

Although there is evidence of atypicalities of social functioning in children with WS, little research has been conducted to directly examine the peer relations of these children. As part of a larger investigation of adaptive functioning, Greer, Brown, Pai, Choudry and Klein (1997) obtained parent ratings of social skills via the Vineland Adaptive Behaviour Scale (4-18 years, N=15; Sparrow, Cicchetti, & Balla, 2005). Parents reported that less than one third of the children with WS had social contacts or engaged in extra-curricular activities with peers. These findings align with previous research reporting that children with WS experience significantly higher levels of social isolation than their typical peers (Udwin et al., 1987; Udwin & Yule, 1991).

Evidence also indicates that peer relations are an area of substantial difficulty for many adults with the disorder (Davies, Udwin, & Howlin, 1998; Elison, Stinton, & Howlin, 2010; Udwin, 1990). Davies and colleagues (1998) conducted interviews with the parents/caregivers of WS adults (N=70) and reported atypical patterns of social interaction. Most WS adults were reported to be socially disinhibited (94%), with characteristic features including asking inappropriate questions (67%) and exhibiting over-tactile behaviour (59%). Crucially, the parents reported that over 95% had experienced friendship difficulties.

Similarly, Elison et al. (2010) revealed that social inclusion was a significant issue for WS adults and under half were said to have any social interests outside the home. Among those who engaged in extra-curricular activities with peers (41%), almost half were reported to struggle to participate. The patterns of peer relation and social interaction difficulties reported in these studies may also relate to increased experiences of bullying among those with WS (Fisher, Lough, Griffin, & Lane, 2017). These studies all contribute to our understanding of peer relations and social interactions in WS adults but understanding these issues in children, and in direct comparison to population norms, has been neglected to date. There is a need for further investigation with more targeted measures probing the nature of friendship experiences to understand how these issues contribute to the broader WS social phenotype and daily lives of children with WS.

The current study is one of the first to use a multi-informant approach to examine the characteristics of peer relations in children with WS across social contexts. In addition to measuring peer relations across standardised measures and in comparison to population norms, this current study shows innovation by using newly developed questionnaires which were designed to tap into context-specific and cross-context features of these peer relationships. While previous research has primarily focused on parents' perspectives, the current study used a multi-informant approach, recruiting both parents and teachers. Incorporating the views of teachers in this research was considered to be important given that peer relations are primarily observed within a school context. Finally, the current study aimed to examine the relationship between atypicalities of social functioning and the quality of peer relations in children with WS which has not been investigated before. It was hypothesised that children with WS would demonstrate impairments across social and communicative domains and greater friendship difficulties than evident from the normative population. It was also hypothesised that there would be a positive association between parents' and teachers'

ratings of peer relation difficulties. Finally, it was expected that there would be a significant positive association between the severity of social and communicative impairment and the degree of peer relationship difficulties (in that children reported to have more significant social impairments would also be reported to show greater difficulty with peer relations).

Method

Participants

Parents (19 mothers and two fathers) of 21 children with WS (12F, 9M) were recruited from the UK. The parents reported on children with WS ranging in age from 7 years 6 months to 16 years 6 months (mean: 11y, 9m; median: 11y, 8m; interquartile range (IQR): 9y, 5m – 14y, 1m). The children with WS scored a mean of 67.05 (SD: 19.52; median score: 60, IQR: 51 – 87.5) in the British Picture Vocabulary Scale (BPVS: Dunn, Dunn, Whetton, & Burley, 1997) measuring verbal performance, and a mean score of 16.67 (SD: 3.79; maximum possible 36; median score: 17; IQR: 13.5 – 18.5) on Ravens Coloured Progressive Matrix (RCPM: Raven, Raven & Court, 1998) measuring visual-spatial performance. Families were members of the Williams Syndrome Foundation UK charity (WSF). All children had previously received a formal diagnosis of WS confirmed through positive genetic fluorescent in situ hybridisation testing. 14 of the children attended a mainstream school and seven attended a specialist provision school. 20 (95%) parents provided permission for the researchers to contact their child's class teacher to seek his or her participation in this research project. 18 of the teachers subsequently agreed to participate.

Measures

The Social Responsiveness Scale, Second Edition – SRS-2 (Constantino & Gruber, 2012)

This is a 65-item questionnaire which assesses social impairments in children aged 4 to 18 years. This questionnaire has been previously administered as a measure of social functioning in children with WS (Klein-Tasman et al., 2011; Lough et al., 2016; Van der Fluit et al., 2012). The scale measures items on five domains: social awareness, social cognition, social communication, social motivation and autism mannerisms/restricted interests and repetitive behaviour. Respondents (parents/caregivers) are asked to read each statement and rate the extent to which the item applies to the child using a 4-point Likert scale ('not true' - 'almost always true'). T-scores are calculated, where the children's raw scores are compared to a normative sample of children of the same gender. According to Constantino and Gruber (2012), a T score of below 60 indicates 'normal' range of social functioning, 60-65 indicates mild impairment, 66-75 moderate impairment and above 75 indicates severe impairment of social interaction. The SRS-2 demonstrated strong internal reliability ($\alpha = .95$) and strong correspondence between informants ($r = .61$) when tested with a large normative sample (Constantino & Gruber, 2012) and showed strong internal reliability for the current sample ($\alpha = .84$).

The Strengths and Difficulties Questionnaire - SDQ (Goodman, 1997)

The SDQ consists of 25 items which measure five constructs of behaviour: emotional symptoms, conduct problems, hyperactivity/inattention, peer problems and prosocial behaviour. Respondents are instructed to read each statement carefully and to rate the extent to which the item applies to the child, using a 3-point Likert scale (0 - 2). Possible scores for each sub-scale range from 0 – 10. Scores from the first four domains are combined to form a composite score termed the 'total difficulties score' (TDS). Possible scores range from 0 – 40, with a score of 17 and above classified as 'abnormal' functioning. The SDQ demonstrates good internal reliability when administered with a normative population sample (Goodman, 2001; $\alpha = .73$). The SDQ demonstrated strong internal reliability when administered with the

parents (α = .80) in the current study, and satisfactory internal reliability when administered with the teachers (α = .64). Published normative data are available for a large child population sample (Meltzer, Gatward, Goodman, & Ford, 2000; N=10,438; age 5-15 years), with ratings from the children's parents (N = 10, 298) and teachers (N = 8, 208).

The Vineland Adaptive Behavior Scales, Second edition – Survey Interview Form - VABS II (Sparrow et al., 2005)

This provides a measure of adaptive behaviour and daily living skills across the lifespan (0-90 years). All items are administered by the researcher using the format of a semi-structured interview. The socialisation construct from the VABS was used to measure the children's social adaptive functioning. This measures interpersonal relationships (38 items), play/leisure time (31 items) and coping skills (30 items). All items are assigned a score of 0, 1 or 2, with higher scores indicating greater social adaptive functioning. An individual's scores across these three subscales are combined to form a domain composite score. Following the guidelines of Sparrow et al. (2005), the composite score is categorised into five classifications of adaptive functioning (high, moderately high, adequate, moderately low and low). Previous researchers reported that the VABS socialisation domain demonstrated high internal reliability with split half reliability scores which ranged between .84 to .93, strong test-retest reliability with correlation values which ranged between .76 to .92, and strong inter-rater reliability with correlation values which ranged between .71 to .81 (Sparrow et al., 2005). This measure showed good internal reliability for the current sample (α = .76). Published normative data are available for a large child population sample of comparable age to the current participant sample (Sparrow et al., 2005; N=1,505, age 7-15 years; 753M, 752F).

Peer interactions questionnaire (parent report)

A six-item scale was developed (see supplementary materials) for this study in response to the lack of appropriate measures of peer relations among children with WS. The questionnaire was designed to measure the children's ability to develop and sustain peer relationships and their social inclusion by peers. Parents are instructed to rate the extent to which each statement applies to the child on a five-point Likert scale, where 1 indicates 'Strongly disagree' and 5 indicates 'Strongly agree'. Item 3 in this scale was reverse scored. Possible total scores range from 6 to 30. A higher score indicates a higher quality of peer relationships. Items in this questionnaire showed strong internal reliability ($\alpha = .78$) and construct validity as indicated by the strong negative association with parents' ratings in the peer problems sub-scale of the SDQ ($r = -.66$, $p = .001$). In order to examine the frequency of the different social experiences measured within this scale (e.g. attendance at birthday parties, frequent fall outs with peers), the Likert scale data were converted to a dichotomous variable for analysis, where the children were assigned a score of '1' where their parent responded 'agree' or 'strongly agree' to the statement, (4 or 5 in the original Likert scale) and a score of '0' where their parent responded 'neither agree nor disagree', 'disagree' or 'strongly disagree' to the statement (1 - 3 in the original Likert scale). Frequencies were computed for each item in the questionnaire.

Peer interactions questionnaire (teacher report)

An 11-item questionnaire was developed (see supplementary materials) for the purpose of this study to measure teachers' perceptions of the peer interactions of the children with WS. This questionnaire was designed to measure the children's ability to develop and sustain friendships with their classmates and social inclusion within school. All items are presented on a five-point Likert scale, where 1 indicates 'strongly disagree' and 5 indicates 'strongly agree'. Items 3, 4, 5, 6 and 10 were reversed scored. Possible total scores range from 11 to 55. A higher score indicates a higher quality of peer relationships. Frequency

statistics were also computed using the method described above. The items demonstrated strong internal reliability (α = .76) when tested with the teachers in the current sample and strong construct validity as indicated by the strong negative association to the teachers' ratings in the peer problems sub-scale of the SDQ (r = -.75, p < .001).

Procedure

Full ethical approval was obtained from the ethics committee at the first author's institution. The children with WS and their parents were visited at home where they participated in all components of the study. Written informed consent was sought from parents and verbal consent was sought from the children. The children with WS completed the BPVS II and RCPM which provided assessments of intellectual functioning. The parents and teachers of the children with WS were invited to complete several questionnaires as detailed above.

Data analysis

The peer relation and socialisation ratings of the children with WS on the standardised measures were compared to normative population statistics (SRS-2, SDQ & VABS II). Bivariate Pearson's correlations examined the relationships between chronological age, verbal and non-verbal ability, indices of social functioning (SRS) and peer relations (SDQ) in children with WS. Bivariate Pearson's correlations were also conducted to examine the association between parents' and teachers' ratings on the SDQ. Cohen's Kappa was calculated to examine the level of agreement between parent and teacher informants on the key overlapping social characteristics (as measured in the novel questionnaires). Across all analyses, one-tailed directional hypotheses were tested with consideration to previous research which provides initial evidence of social difficulties in this population, allowing

specific predictions to be tested in the current study (Greer et al., 1997; Klein-Tasman et al., 2011).

Results

Comparing the children with WS with normative population scores on standardised measures of social skills

Profile of social functioning

As demonstrated in Figure 1, 38% of the children with WS were reported to show moderate levels of impairment in everyday reciprocal social interactions, and 48% were reported to show severe levels of impairment, as measured in the SRS 2. Only 10% of the children with WS scored within the normal range of social functioning. The majority of the children were classified as being severely impaired within the social awareness (52%), social cognition (67%) and autism mannerisms (76%) sub-scales and moderately impaired on the social communication sub-scale (57%). Fewer children were classified within the moderate (29%) and severe (10%) impairment categories on the social motivation domain.

--- Insert Figure 1 ---

Peer relations

Parents and teachers rated the children with WS on the SDQ, providing a measure of everyday functioning and an assessment of peer relations (Table 1).

--- Insert Table 1 ---

Children with WS were reported by their parents and teachers to have substantially more problems within peer relations compared to the normative population. One-sample t-tests (against the population mean) showed that compared to population norms the WS group were

rated significantly higher by their parents on the total difficulties composite scale (t (20) = 8.75, $p<.001$, $d = 1.92$) and peer problems sub-scale (t (20) = 7.77, $p<.001$, $d = 1.84$). Children with WS were also rated significantly higher than population norms by teachers on the total difficulties composite scale (t (17) = 8.24, $p<.001$, $d = 1.72$) and peer problems sub-scale (t (17) = 4.42, $p<.001$, $d = 1.19$). It is also worth noting the vast heterogeneity seen in Table 1.

Bivariate Pearson's correlations were conducted to examine the association between parents' and teachers' ratings on the SDQ showing a significant moderate positive association between parents' and teachers' scores on the total difficulties composite scale (r=.55, $p=.009$) and peer-problems sub-scale (r=.46, $p=.029$).

Socialisation skills

Parents rated the children with WS on the socialisation domain of the VABS (Table 2).

--- Insert Table 2 ---

Table 2 indicates that the children with WS demonstrate socialisation skills that are substantially below the normative population. Across all sub-scales the children with WS performed significantly lower than would be expected for their age, with the mean age equivalent for each sub-scale ranging from 5- to 7-years (interpersonal relationships: 7y, 9m; play and leisure time: 5y 5m; coping skills: 6y, 1m), substantially lower than the mean chronological age of the sample (M = 11y, 9m). The frequency of children with WS classified, according to the guidelines of Sparrow et al. (2005), in the low, moderately low, adequate, moderately high and high range of adaptive functioning are illustrated in Figure 2. Frequencies are reported for each of the socialisation sub-scales and the socialisation domain composite score.

--- Insert Figure 2 ---

Independent samples t-tests compared parent ratings to a normative child population sample (Sparrow et al., 2005) on the VABS socialisation scale. The children with WS were rated significantly poorer than the normative population sample on interpersonal relationships (t (20) = -3.10, p = .003; d = 0.89), play and leisure time (t (20) = -5.62, p <.001; d = 1.81), coping skills (t (20) = -5.32, p <.001; d = 1.25) and for the overall standardised socialisation score (t (20) = -7.17, p< .001; d = 1.71).

Associations between age, intellectual ability, atypicalities of social functioning and peer relationship difficulties in children with WS

Bivariate Pearson's correlations were conducted (Table 3) to examine the relationships between chronological age, verbal and non-verbal ability, social functioning impairments (SRS-2) and peer relationships difficulties (SDQ).

---Insert Table 3 ---

Chronological age and intellectual ability (verbal and non-verbal skills) were not significantly associated with parents' reports of social skills, or with parent or teacher ratings of peer relationship difficulties. There was a moderate positive association between social functioning impairments on the SRS-2 and peer relationship difficulties as rated by parents (r = .44) and teachers (r = .44) on the SDQ.

Examining the characteristics of peer relations across social contexts

Mean score on the novel parent rated peer interaction questionnaire was 18.71 (SD 5.65; range 8-29; possible range 6-30; higher score indicates higher quality peer relations). Mean score on the novel teacher rated peer interaction questionnaire was 37.28 (SD 6.79;

range 26-52; possible range 11-55; higher score indicates higher quality peer relations). There was within-syndrome variability reported for the quality of the children's peer relations.

For each of the social characteristics measured within the novel questionnaires, frequencies are reported on the proportion of the sample for whom that statement is true of (see method section for further details on scoring). Within parent and teacher questionnaires, several items were included which were applicable across both social contexts (e.g. ability to sustain friendships). Table 4 reports frequency statistics for these key overlapping characteristics. Cohen's Kappa was calculated to examine the level of inter-rater agreement between parents and teachers on each of these overlapping characteristics. Parents and teachers showed no significant agreement on items assessing the presence of a friendship (k = .008, 95% CI, -0.24 to 0.26, p = .947) and the ability to sustain friendships (k = -.147, 95% CI, -0.39 to 0.09, p = .223). Parents and teachers showed slight agreement on their ratings of the children's experience of falling outs with peers (k = .274, 95% CI, 0.004 to 0.54, p = .020).

-- Insert Table 4 ---

Additional items were incorporated to examine context-specific features of peer relations, providing an assessment of peer relationships within the home and school (Table 5).

-- Insert Table 5 ---

Social exclusion was reported to be an area of difficulty for the majority of the children with WS. These social difficulties were evident within school, with regard to the children's inclusion by peers during recreational breaks and within classroom activities, and outside school, with less than half of the children reported to be invited to their peers' homes or birthday parties. This consistency across parent and teacher reports suggests that while

occurring in different forms, social exclusion was problematic across both the home and school environment. Despite many of the children with WS being reported to show difficulties sustaining friendships and often experiencing exclusion from peer activities, none of the children were picked on by their peers and most were considered to be well-liked by their peers according to teacher report.

Discussion

The current findings provide preliminary evidence of substantial social difficulties in children with WS across standardised measures when compared to normative child population samples. The study examined the characteristics of these peer relationship difficulties in depth, using a multi-informant approach to examine across social contexts. The findings indicate atypicalities of social functioning, difficulties developing and sustaining reciprocal peer relations and low levels of inclusion by peers. These social difficulties were evident across social contexts. Atypicalities of social and communicative functioning showed a moderate correlation with the quality of the children's reported peer relationships. This study builds on previous research by obtaining a more detailed investigation of the social functioning and peer relations of children with WS across social settings.

Atypicalities of social functioning

The majority of the children with WS demonstrated moderate or severe impairments of social functioning on the standardised SRS-2. These findings align with previous research (Klein-Tasman et al., 2011; Lough et al., 2016). Relatively fewer children with WS were classified as moderately or severely impaired within the social motivation domain. However, the SRS only captures one aspect of social motivation and focuses on social withdrawal. While the children with WS do not show difficulties in social withdrawal, these children often show atypical patterns of social motivation as highlighted by a heightened interest to

approach and engage in social interactions (Bellugi et al., 1999; Dodd, Porter, Peters, & Rapee, 2010). Consequently, the children's social motivation scores in this scale should be interpreted with caution (see Riby et al., 2017 for a full discussion on the issue of capturing atypical social motivation in WS from the SRS measure).

Social cognition was identified as an area of particular difficulty. The current questionnaire findings provide support for previous experimental research which identified substantial theory of mind difficulties (Porter et al., 2008; Tager-Flusberg & Sullivan, 2000). These atypicalities of social cognition were reported to be shown through the children's difficulties interpreting facial expressions and tone of voice, and their difficulties engaging in pretend play. These impairments across aspects of social and communicative functioning may hinder the children's ability to engage in reciprocal social interactions with their peers.

Peer relationship difficulties

Peer relationship difficulties were prevalent within this sample. Children with WS scored significantly lower than a normative child population sample (Sparrow et al., 2005) on the socialisation domain of the VABS, in line with previous work (Greer et al., 1997). However, the current study builds upon previous research by analysing scores at both the domain and sub-domain level. The children with WS scored significantly poorer than the normative population across the VABS sub-scales of interpersonal relations, play/leisure skills and coping skills.

Previous research examining peer relations in WS has predominantly used parent ratings (Elison et al., 2010; Greer et al., 1997). The present study builds on this, using amulti-informant approach to include both parents and teachers. The school environment provides a context for children to interact with their peers and can be suggested to provide an optimal setting to assess peer interactions (Wentzel & Miele, 2016). Children with WS were rated

significantly higher in peer problems within the SDQ than the normative population across both parent and teacher reports. There was a moderate positive association between parents' and teachers' ratings of peer problems. This suggests that these social difficulties transcend across social settings but that some differences between informants do occur, with parents rating the children slightly higher in peer problems on average than the teachers.

While a small number of research studies have been conducted to examine experiences of social isolation in this population (Gosch & Pankau, 1994; Greer et al., 1997), the current study is one of the first to examine the characteristics of peer relationships among children with WS and how these social experiences are displayed across social contexts. Difficulties identified in the current study included the absence of a close friendship, difficulties developing and sustaining peer relationships and low levels of social inclusion by peers. Despite these peer relationship difficulties, very few children were reported to have falling outs with peers and none of the children were reported to be bullied by their peers. These findings contrast with previous evidence of peer victimisation among adolescents with intellectual disabilities (Zeedyk, Rodriguez, Tipton, Baker, & Blacher, 2014) and autistic adolescents (Fisher & Taylor, 2016). This low level of peer conflict, a positive trait in friendship development, is an interesting avenue to explore within future research. In particular, there is a need to examine the syndrome specificity of these experiences, and to consider this within the context of the broader WS social phenotype.

Although peer difficulties were evident across parent and teacher ratings, as indicated on the SDQ, interestingly, some differences within observations were revealed when parents and teachers were asked to rate specific characteristics of these peer relationships, including the ability to sustain friendships. This indicates the value of including both parent and teacher informants in research, with peer interactions potentially manifesting differently across these social contexts. However, it should be noted that no information was collected on the

teachers' level of experience or familiarity with the child. Differences in the informants' familiarity with the child with WS could provide a potential explanation for the low level of agreement between parents and teachers in their perceptions of these characteristics of the child's peer relationships. Further research is required, with a larger sample where possible and validated measures, to further develop our understanding of these cross-context similarities and differences, controlling for the socio-demographic characteristics of parents and teachers.

While the current findings indicate substantial peer relationship difficulties in WS, there was within-syndrome variability and the findings build on current evidence of within-syndrome variance in other aspects of the WS profile such as cognition (Little et al., 2013; Lough et al., 2016). Given the relatively large age range and spectrum of intellectual abilities shown in the current participant sample, future research should seek to recruit a more select sample with regards to age and ability levels when examining the peer relations of children with WS.

The role of social functioning skills in the quality of peer relations in children with WS

There was no significant association between chronological age, intellectual functioning and the quality of peer relations. This suggests that the peer relationship difficulties are stable across this period of development and indeed the literature involving WS adults has reported similar difficulties extending into adulthood. Furthermore, the current study reported a moderate positive association between atypicalities of social functioning and greater peer problems suggesting that children with WS may experience difficulties in developing and sustaining peer relationships arising from impairments across different aspects of social functioning (which we also know to be evident across the developmental spectrum). Social difficulties that might feed into peer relation difficulties include

impairments detecting and interpreting social cues (mapping to numerous previous studies such as Lough et al., 2015; Porter et al., 2008; Tager-Flusberg & Sullivan, 2000). Further research understanding the social mechanisms underlying peer relation difficulties is important and the current study begins to delve into this issue.

Limitations and future research

While the current research study has provided important novel insights into the peer relations of children with WS, we must acknowledge limitations and caveats to be considered in the interpretation of the findings and their implications. Firstly, the current analyses and interpretations are based on a relatively small sample size of 21 children with WS. This sample size is completely in line with previous research arising from the rarity of this disorder and the size of the UK where the research was conducted (Järvinen-Pasley et al., 2010). This sample size posed implications for the analysis, where it was not possible to draw conclusions regarding the predictive relationship between social functioning impairments and peer relationship quality due to a lack of statistical power. The small sample size also made it difficult to draw strong comparisons between the peer relationships of the children with WS who attended a mainstream school and those who attended a specialist provision school. Within future research, obtaining a larger sample size would allow investigation into whether the characteristics of a child's peer group have an impact on the child's peer relationships.

There were no normative data for the parent and teacher 'peer interaction questionnaire'. This makes it difficult to draw strong conclusions regarding the atypical nature of these peer interactions in WS. Within future research, more extensive investigations could be achieved by drawing comparisons between the characteristics of peer interactions in children with WS, typically developing children and other developmental disorder groups. This would provide insight into the atypical nature of these peer interactions among children

with WS and the syndrome-specificity of the social difficulties. Finally, it should be noted that the teachers in the current study did not complete the SRS, therefore cross-informant comparisons were not possible on this measure. However, previous research has indicated that parents and teachers showed strong concordance on this measure (Klein-Tasman et al., 2011).

Conclusion

The current study provides preliminary evidence for substantial atypicalities within social functioning and peer interactions in children with WS. Children with WS scored significantly lower than the normative population across standardised measures of social functioning, with these difficulties present across parent and teacher reports. The use of specifically designed questionnaires provided further insight into the atypical nature of the children's peer relationships and exclusion from their peer group. Impairments in social functioning were identified as strong underlying factors in the children's peer relationship difficulties. It will be important, in future research, to determine the replicability of the present results among samples from different communities and with different levels of educational support. These findings may be useful in informing the design of future interventions to target these areas of social difficulty.

Compliance with ethical standards

Conflict of Interest: The authors declare that they have no conflict of interest.

Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent: Informed consent was obtained from all individual participants included in the study.

Author note

Amanda E. Gillooly, School of Psychological Sciences and Health, University of Strathclyde, 40 George Street, G1 1QE, Scotland, UK

Deborah M. Riby, Department of Psychology, Durham University, South Road, Durham, DH1 3LE, England, UK

Kevin Durkin, School of Psychological Sciences and Health, University of Strathclyde, 40 George Street, G1 1QE, Scotland, UK

Sinéad M. Rhodes, Salvesen Mindroom Research Centre, Child Life & Health, Centre for Clinical Brain Sciences, University of Edinburgh, 20 Sylvan Place, Edinburgh, EH9 IUW, Scotland, UK[4]

Amanda E Gillooly is now at the Institute of Health and Wellbeing at the University of Glasgow, Glasgow, Scotland.

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Correspondence concerning this article should be addressed to Sinéad M. Rhodes, Salvesen Mindroom Research Centre, Child Life & Health, Centre for Clinical Brain Sciences, University of Edinburgh, 20 Sylvan Place, Edinburgh, EH9 IUW, Scotland, UK.

sinead.rhodes@ed.ac.uk, 0131 536 0642

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Table 1

Parent and teacher ratings of SDQ Total difficulties and SDQ Peer problems for children with WS compared to normative population sample

		WS (N=21)		Normative population	
		Mean (SD)	Range[a]	Mean (SD)	
Parent ratings	Total Diff[b]	19.62 (5.88)	7 – 28	8.4 (5.8)	p < .001
	Peer problems	4.95 (2.04)	0 – 7	1.5 (1.7)	p < .001
Teacher ratings	Total Diff[a]	16.00 (4.84)	5 – 24	6.6 (6.0)	p < .001
	Peer problems	4.00 (2.50)	0 – 9	1.4 (1.8)	p < .001

Note. All scores reported are raw scores

[a] Range of scores for the normative population were not available

[b] Total difficulties composite score

Table 2

VABS socialisation ratings for children with WS compared to normative population sample

	WS (N=21)		Normative population	
	Mean (SD)	Range	Mean (SD)	
Interpersonal relationships (raw)	57.43 (12.82)	34 – 80	66.09 (6.67)	p = .003
Play and leisure time (raw)	34.05 (15.68)	8 – 67	53.27 (5.59)	p < .001
Coping skills (raw)	31.48 (12.16)	9 – 54	45.59 (10.47)	p < .001
Socialisation (standardised)	73.38 (17.30)	50 – 119	100.45 (14.4)	p < .001

Table 3

Pearson's bivariate correlations between age, verbal and non-verbal ability, social functioning impairments (SRS-2) and parent and teacher ratings of peer relations in children with WS (N=21)

	1	2	3	4	5	6
Age		.70***	.53**	-.12	.14	-.24
Verbal ability (BPVS)			.58**	-.23	.32	.05
Non-verbal ability (RCPM)				.04	.21	.18
SRS total raw score					.44*	.44*
SDQ peer problems (parent report)						.46*
SDQ peer problems (teacher report)						

*$p < .05$, ** $p < .01$, *** $p < .001$

Table 4

Comparing parent and teacher reports on the quality of peer relations in children with WS

	Parents (N=21)	Teachers (N=18)	Cohen's K
One or more friends	9 (42.9%)	8 (44.4%)	k = .008, *p* = .947
Ability to sustain friendships	10 (47.6%)	8 (44.4%)	k = -.147, *p* = .223
Experiences falling-outs with peers	5 (23.8%)	4 (22.2%)	k = .274, *p* = .020

Table 5

Examining context specific characteristics of peer relations among children with WS in the home and school environment

	Item	Frequency
Parents (N=21)	Invited to peers' homes	8 (38.1%)
	Invited to peers' birthday parties	10 (47.6%)
	Attends extra-curricular activities with peers	13 (61.9%)
Teachers (N=18)	Included by peers during recreational breaks	7 (38.9%)
	Included by peers during conversations in the class	7 (38.9%)
	Chosen as a partner for work activities/P. E	5 (27.8%)
	Greeted by other children around the school	14 (77.8%)
	Picked on by peers	0 (0%)

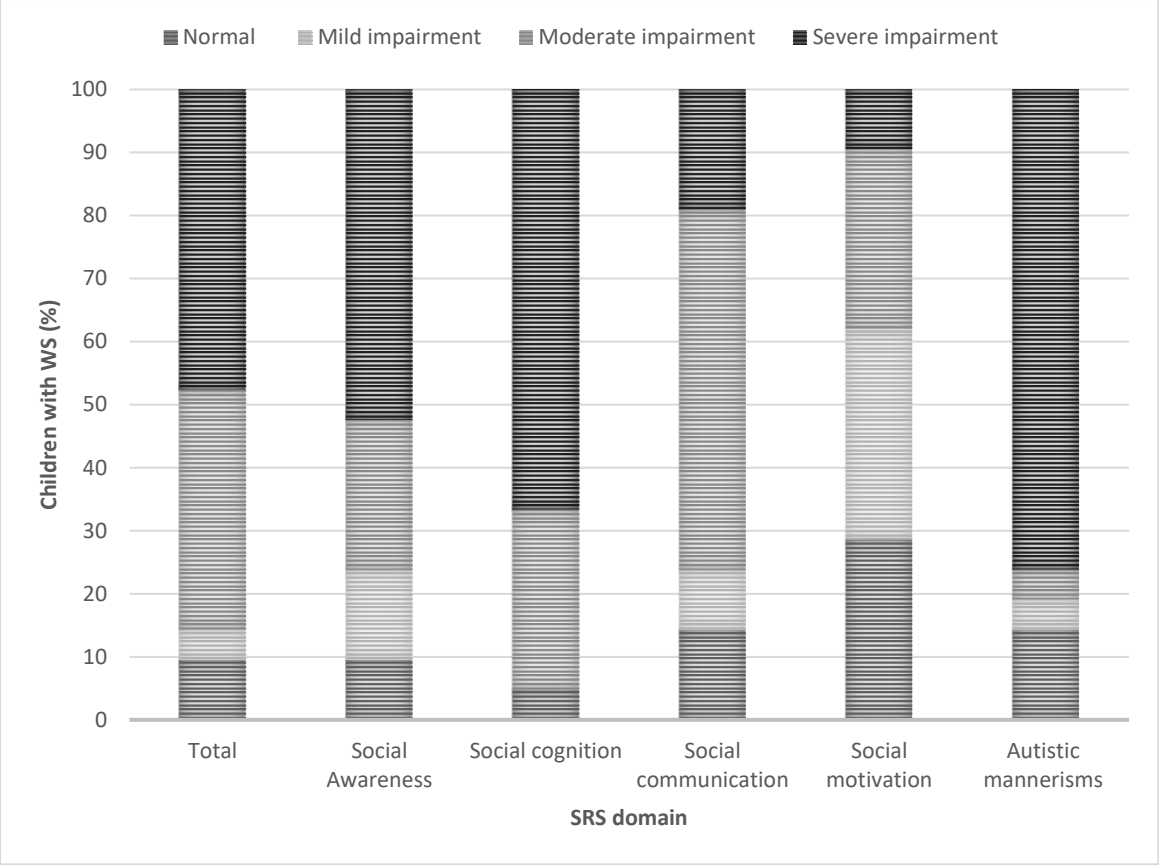

Fig.1 Distribution of classifications for the sub-scale and total score in the SRS-2 among the children with WS (N=21)

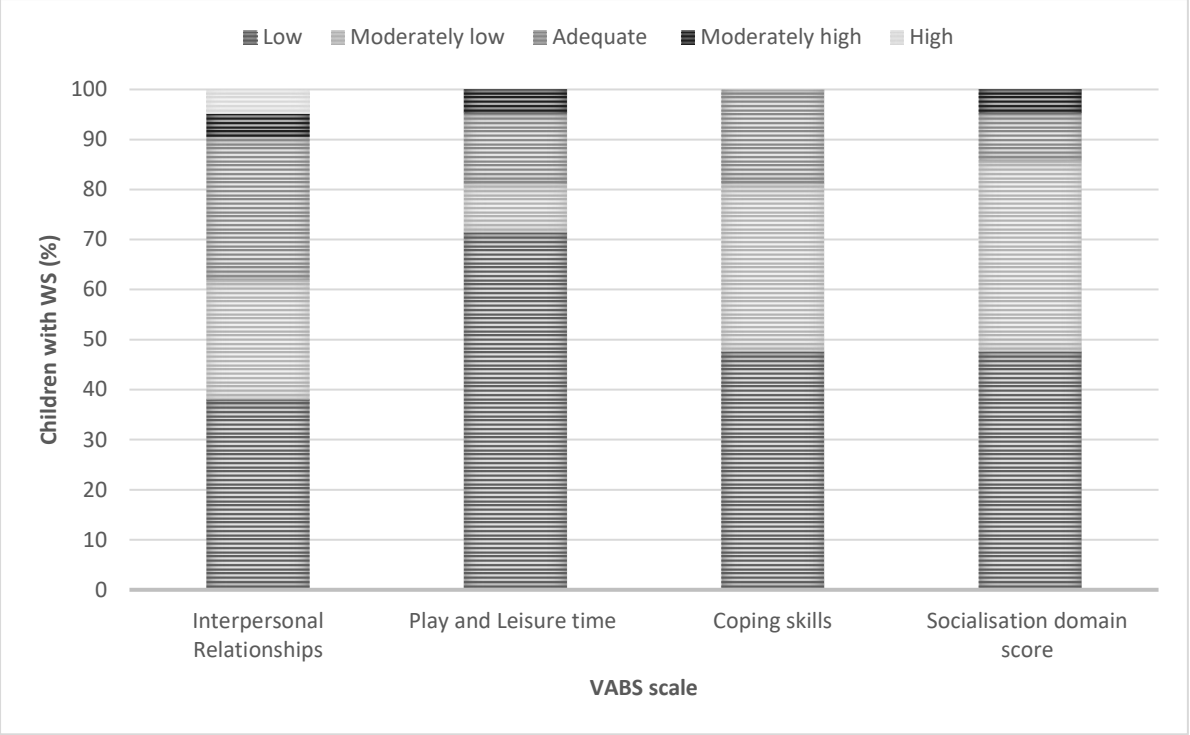

Fig.2 Distribution of classification (level of ability) of adaptive functioning on the VABS socialisation domain among the children with WS (N=21)